Impact Peak, Knee Muscle Cocontraction, and Knee Angle during Early Contact of Running in Children with Diplegic Cerebral Palsy

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By

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DEDICATIONS

To my husband, Harris

in deep appreciation of your understanding, support and encouragement.

To my beloved parents,

in gratitude for your love and endless support.

To devoted and respected teachers,

Specially Dr. Margo Orlin

in gratefulness for teaching me to become who I am.

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ABSTRACT

Impact Peak, Knee Muscle Cocontraction, and Knee Angle during Early Contact of Running in Children with Diplegic Cerebral Palsy

Sirinart Laibsirinon

Purpose: Some children with ambulatory diplegic cerebral palsy (CP) are able to run. Running can enjoyable and a simple form of exercise for children with CP to stay physically active. However, they may be at risk for lower extremity injuries due to atypically high impact force. Excessive knee muscle cocontraction and the inability to modify knee angle during early contact phase of running may contribute to ineffective force attenuation, resulting in atypical high impact peak and load rates during running. The purpose of this study was to compare impact peak, load rates, knee stiffness, knee muscle cocontraction and knee contact angle during early stance phase of running between children with CP and children with typical development (TD).

Method: Six children with CP and eight with TD participated in this study. Because of difference in strike patterns during running in children with CP that had not been anticipated in the initial research plan. Therefore, kinetic and knee angle data of four identified children with TD were included from unpublished database and were used to compare those with children with CP who contacted the ground with midfoot and rearfoot from this study. After static trials were collected in all children, children ran at a comfortable speed in the motion analysis laboratory equipped with eight video-camera, three force plates and a wireless surface electromyography (EMG). Three acceptable trials of running in which leg hitting the middle of the force plates were required for data analysis. Kinetics and knee contact angle were post-processed, analyzed and computed at

a period of initial contact (IC) to impact peak (IP) of vertical ground reaction force (GRF). Impact peak, average and instantaneous load rates, knee stiffness, knee muscle cocontraction, and knee contact angle at IC and at IP as well as knee excursion from IC to IP were dependent variables of this study. Mann-Whitney *U* test were used to compare dependent variables between children with CP and children with TD. Effect sizes (ES) of *r* of all variables were also reported.

Results: Impact force and load rates were significantly higher in the more involved and the less involved legs of children with CP, compared to those with TD although children with CP and children with TD ran at similar running speeds. No other variables were different between the two groups.

Conclusion: Due to very small sample size, the findings from this study still need further investigation. The key finding suggests that children with CP have atypically high impact peak and load rates despite slow running speeds, when compared with children with TD. High impact peak and load rates may be used to determine children with CP who may be at risk for lower extremity injuries. Parents, teachers and health professionals such as physical therapists, who work with children with CP should consider atypically high impact force and load rates if children demonstrate any pain and/or musculoskelatal discomfort during and after running. Proper footwear, orthoses, and running retraining have been reported to reduce impact forces and load rates in runners without neuromuscular disorders, which could be potential options to minimize the risk for lower extremity injuries in children with CP. However, these interventions need to be investigated in children with CP.

CHAPTER 1

Research Proposal

1.1 Specific Aims

Running is an enjoyable form of physical activity which can improve muscular and cardiovascular fitness in young children. Children run to participate in recreational and sport activities. Children with spastic diplegic cerebral palsy (CP) who are ambulatory show desire to run and some run often during play with peers and siblings. Unfortunately, running has not typically been a part of a physical therapy examination for children with CP. This is, in part, because participation of children with CP, in sport, recreation and physical activities that include running are not formally promoted. This may be attributed to fear of developing injuries that may result from running. This fear may be due to the lack of empirical knowledge regarding actual causes or risk factors contributing to injuries due to running in children with CP.

In addition to training errors and lower extremity malalignment, biomechanical variables have been speculated to be primary causes of running injuries (Malisoux, Nielsen, Urhausen, & Theisen, 2014; Messier et al., 2008). Some are correlated with overuse running injuries which include atypically high vertical impact peak and high load rate, and excessive leg and knee joint stiffness (Hreljac, 2005; Milner, Ferber, Pollard, Hamill, & Davis, 2006). In non-injured runners, increased impact and loads are attenuated into optimal level to avoid injuries. Kinematic adaptation of lower extremities has been suggested to be one of several strategies to attenuate forces. Increased knee flexion at early contact has been suggested to be a primary kinematic adaptation (Derrick, 2004; Hreljac, 2004, 2005; McMahon, Valiant, & Frederic, 1987). More knee flexion at early contact phase of running can decrease impact forces and consequently reduce injury potential.

The successful adaptation of knee flexion is regulated by normal neuromuscular control. During high loading movement, such as running, muscle cocontraction around the knee joint is an important part of normal neuromuscular control. Cocontraction serves as a supplement to joint ligaments in order to maintain joint stability and prevent injury (Noyes, Barber, & Mooar, 1989; Woo, Hollis, Adams, Lyon, & Takai, 1991). However, amounts of muscle cocontraction must be optimal. Excessive muscle cocontraction may cause a stiffer joint and leads to failure to modify knee flexion angle at early contact phase, resulting in abnormally high impact force. Optimal muscle cocontraction should typically occur in individuals without neuromuscular disorders, unlike children with CP who have impaired neuromuscular control. Excessive cocontraction of knee muscle has been reported in children with CP during walking but yet during running (Keefer et al., 2004; Unnithan, Dowling, Frost, & Oded, 1996).

In children with CP, excessive knee muscle cocontraction and difficulty in modifying the sagittal knee angle may lead to excessive impact force, load rate and knee joint stiffness during early contact phase of running. Therefore, the fear of developing injuries due to running in children with CP may be a reasonable one if children with CP are not able to optimize impact forces into non-injury zone. This may cause concern for ambulatory children with CP who desire to run. The inability to attenuate high impact forces over time during running may be a predisposing factor to the development of musculoskeletal joint trauma. The concern is the potential long term impact if there is a lack of force attenuation on lower extremity joints. There are critical gaps in this research area. Little is known about the behavior of biomechanical factors including impact force, load rate and knee joint stiffness, knee muscle cocontraction and knee contact angle during early stance of running in children with CP. Only one study reported a trend of higher impact force, load rate and leg stiffness during running in children with CP, compared with children with typical development (TD) (Laibsirinon & Orlin, 2010; Orlin & Laibsirinon, 2010). Another study reported increased knee flexion at initial contact but decreased knee flexion excursion during an entire contact phase in children with CP, compared with those without CP (Davids, Bagley, & Bryan, 1998). This indicates that although knee flexion is present, the knee joint is stiff and does not flex and extend, as it should during the running cycle. Impact force and load rate, knee joint stiffness and knee angle as well as muscle cocontraction, that may be used to identify risk for running related injuries have not been systematically investigated in children with CP. Lack of this knowledge limits health professionals from identifying which children may be at risk for musculoskeletal injuries sustained during running. Knowing these kinetics, knee angle and knee muscle cocontraction provide clinical rationale for the inclusion of examination procedures and intervention to prevent potential injuries. This knowledge can be used to ensure safe running in children with ambulatory CP. A conceptual framework as shown in Figure 1.2 has been formulated to systematically characterize biomechanical factors and knee muscle cocontraction during running in children.

The ***long term goal*** of this research is the development of comprehensive examination and intervention protocols to avoid possible injuries and enhance running for ambulatory children with diplegic CP. The ***objective of this dissertation proposal*** is to compare kinetic variables and knee flexion angle as well as knee muscle cocontraction between ambulatory children with diplegic CP and children with TD during the period of initial contact to impact peak. The ***central hypothesis*** is that children with diplegic CP will demonstrate higher impact force, higher load rate and higher knee joint stiffness during period of initial contact to impact peak due to the inability to adapt knee flexion angle and excessive muscle cocontraction during running. Thus, knee flexion angle at impact peak and knee excursion from initial contact to impact peak as well as knee muscle cocontraction of children with diplegic CP should differ from children with TD. To accomplish the objectives, a specific aim is proposed:

Specific Aim 1: To compare kinetic variables related to running injures, as well as knee flexion angle and knee muscle cocontraction between children with diplegic cerebral palsy (CP) and children with typical development (TD)

Hypothesis 1: Children with CP will demonstrate higher impact peak, load rates and sagittal knee stiffness at a period of initial contact to impact peak, compared to children with TD

Hypothesis 2: Children with CP will demonstrate different knee flexion angle at initial contact and at impact peak, and smaller knee flexion excursion from initial contact to impact peak, compared to children with TD

Hypothesis 3: Children with CP will demonstrate higher knee muscle -cocontraction, compared to children with TD

This research is innovative because it is the first to apply biomechanical factors, shown to be correlated with running injuries in adults, to determine children with CP who are at risk for injury from running. Knowledge of impact force, knee angle and muscle cocontraction will partially explain the biomechanical control of knee joint during running in children with diplegic CP. The expected outcome of this study is that it will be used to inform an inclusion of running-related examination procedures in children with ambulatory CP who desire to run and to provide some justifications for interventions to minimize potential running-related injuries and to maximize running ability in children with CP.

1.2 Significance and Innovation

Increased physical activity and participation in exercise, sport and recreation activities have been widely promoted in children with physical disability including those with CP (Fowler et al., 2007; Murphy & Carbone, 2008; Shikako-Thomas, Kolehmainen, Ketelaar, Bult, & Law, 2014; Verschuren, Darrah, Novak, Ketelaar, & Wiart, 2014). Many researchers and educators advocate that more studies are needed to learn how participation in exercises and sport should be recommended (Fowler et al., 2007; Patel & Greydanus, 2002; Rimmer & Rowland, 2008). Increased physical activity and participation in appropriate types and levels of exercises can improve not only physical fitness, mental health and overall well-being but also can prevent secondary conditions in children with CP (Biddle & Asare, 2011; Johnson, 2009; Thorpe, 2009). Most children with spastic diplegic CP have abilities to walk and to run. Running is economical and universal. Running is a simple form of exercise and physical activities that can be possibly performed in children with ambulatory diplegic CP classified at level I and II of Gross Motor Functional Classification system (GMFCS) (Brunton & Bartlett, 2010; Maher, Williams, Olds, & Lane, 2007; Palisano et al., 1997). Fear of developing injuries and complications as well as lack of information and knowledge are self-identified as barriers to attend physical activity in young adults with physical disabilities, including CP (Buffart, Westendorp, van den Berg-Emons, Stam, & Roebroeck, 2009; Verschuren, Wiart, Hermans, & Ketelaar, 2012). The fear of developing injuries due to running sounds logical and empirical. Unfortunately, much is unknown that limits clear answers for this concern. Biomechanically, greater impact force and higher load rate during running is reported, compared with those during walking (Hreljac, 2004). A number of evidence reveals a strong association of high impact force and load rate with overused injuries in adolescents and adults without neuromuscular disorders (Grimston, Engsberg, Kloiber, & Hanley, 1991; Grimston, Nigg, & Fisher, 1993; Hreljac, 2005; Milner et al., 2006). The critical knowledge of kinetics related to injuries such as impact force, load rate, joint stiffness and kinematics of children with CP during running has been little investigated and reported (Laibsirinon & Orlin, 2010; Orlin & Laibsirinon, 2010). Knee muscle cocontraction which is critical neuromuscular control of joint movement during running is unknown in children with CP. This study will begin to investigate kinetics related to injuries as well as and knee flexion angle and knee muscle cocontraction during running in children with diplegic CP. These knowledges are very significant prior to encourage children with CP to participate in running activities. Knowledge of kinetics, knee flexion angle and knee muscle cocontraction during running will be used to partly inform clinical guideline and clinical decision making to determine risk due to running and warrant safe participation in running in children with CP.

1.3 Background

1.3.1 Cerebral Palsy (CP)

The most widely well-known definition of Cerebral Palsy was reported by Rosenbaum et al in 2007 (Rosenbaum, Paneth, Leviton, Qoldstein, & Bax, 2007). CP is defined as *"a group of permanent disorders of movement and posture, causing activity limitation, that are attributed to non-progressive disturbance that occurs in the developing fetal or infant brain" p.9*. CP is the most common cause of severe physical disability in childhood (Shevell, Dagenais, & Oskoui, 2013). Recent systematic review reported the worldwide prevalence of CP from 1985 to 2004 ranged from 1.10 to 3.6 per 1000 live births (Donald, Samia, Kakooza-Mwesige, & Bearden, 2014; Oskoui, Coutinho, Dykeman, Jetté, & Pringsheim, 2013). The prevalence of CP in USA was reported higher than global record but remained relatively constant from 1996 to 2008, in the approximation of 3.1-3.6 per 1000 live births (Arneson et al., 2009; Christensen et al., 2014; Kirby et al., 2011; Yeargin-Allsopp et al., 2008). About 764,000 children and adults in the United State have one or more of the symptoms of CP (United Cerebral Palsy Organization's fact sheet, 2012). According to RIT International (Research Triangle Park) and CDC, the estimated lifetime cost of persons with CP in the U.S. in 2003 was $11.5 billion and average cost per person was $921,000 (Honeycut et al., 2004). Spastic diplegia is the first or second most common type of limb distribution classification of children with CP in the U.S. and Canada (Gorter et al., 2004; Yeargin-Allsopp et al., 2008). The majority of children with diplegic CP have motor skills classified at Gross Motor Function Classification System (GMFCS) level I which indicated that they can independently walk and run (Gorter et al., 2004). Lower levels of physical activity and physical fitness have been widely reported in children with CP, compared with children without CP (Carlon, Taylor, Dodd, & Shields, 2013; Van Den Berg-Emons et al., 1995). Improving participation in regular physical activity and appropriate exercise program, such as brisk walking or jogging and running since in young age of individuals with CP may maintain functional activities, prevent secondary conditions and possible decline in ambulation (Verschuren et al., 2014).

1.3.2 Risk Factors for Running Related Overuse Injuries at the Knee in Adolescents and Adults without Neuromuscular Disorders

Running is one of the most popular forms of exercise due to health benefits, low cost and convenience. However, injuries are very common. The number of injuries has increased with the increasing popularity of running in the past 3 decades in the Unites State and around the world. The estimated incidence rate during any 1-year period is 37% to 79%, varied between recreational and competitive runners as well as definitions of injury (Lun, Meeuwisse, Stergiou, & Stefanyshyn, 2004; van Gent et al., 2007; van Mechelen, 1992). The most common injuries related to running occur at lower extremity, with predominance for the knee (Hoch, Pepper, & Akuthota, 2005; Taunton et al., 2003; Taunton, Ryan, Clement, McKenzie, & et al., 2002; van Gent et al., 2007; Wen, 2007). The vast majority of injuries is considered "overuse". *Overuse injuries* has been defined as *"an overload of the musculoskelatal structures, resulting from repetitive microtrauma over a period of time without a sole and identifiable event"* (Hreljac, 2005; Saragiotto et al., 2014). Some of the most common clinical conditions that are often considered overuse running injuries at the knee and lower legs include patellofemoral knee pain, shin splints, iliotibial band syndrome and tibial stress fracture (van Mechelen, 1992). Associations between risk factors and overall running injuries have been investigated and reported. The risk factors for overuse running injuries are multifactorial in nature. The variables that have been identified as potential risk factors for overuse injuries at the knee and lower leg in running are commonly divided into 4 main categories which consist of *personal characteristics (eg. gender, age and body mass index), training errors, anatomic and biomechanical variables* (Hreljac, 2005; Magness, Ambegaonkar, Jones, & Caswell, 2011; Wen, 2007). Recent literature reviews and systematic reviews provide a summary of risk factors and associated injuries. These reviews included studies with retrospective, prospective and cross sectional designs in adolescents and adults who are either recreational or competitive runners ((Ferber, Hreljac, & Kendall, 2009; Malisoux et al., 2014; Saragiotto et al., 2014; Wen, 2007; Wen, Puffer, & Schmalzried, 1998; Zadpoor & Nikooyan, 2011).

Personal characteristics that are commonly suspected to be risk factors consist of age, gender and body mass index. Most of review studies revealed no associations between age, gender and running injuries (Saragiotto et al., 2014; D. Wen, 2007). However, increased age, approximately older than 30 years old in both genders, has been associated with tibial stress fracture, patellofemoral pain and iliotibial band syndrome. (J. Taunton et al., 2003; J. E. Taunton et al., 2002). Female gender is reported to experience higher rate of tibial stress fracture than male (J. E. Taunton et al., 2002; D. Wen, 2007). Lower BMI, specifically <25 $kg/m^{2,}$ is found to be at higher risk for tibial stress fracture (Malisoux et al., 2014; J. E. Taunton et al., 2002).

Many training variables which has long been reported to be associated with injuries are *training errors, running surface and running footwear* (Hreljac, 2005; Hreljac, Marshall, & Hume, 2000; Nielsen, Buist, Sørensen, Lind, & Rasmussen, 2012). *Training errors* are concerned with high training variables, fewer rest days and rapid increase in duration or intensity. Confusing and inconsistent definitions of training variables have been used across studies including *volume* (the average mile or kilometers/week*)*, *duration* (average hours or minutes/week or per day), *distance* (miles per week), *frequency* (number of days or times per week) and *intensity* (during training: minute/mile or per km). They are highly recognized as risk factors by clinicians, researchers and running coaches (Hreljac, 2005; Hreljac et al., 2000; Nielsen et al., 2012). A recent systematic review on a relationship between training errors and running related injuries suggested that an increase in any training variables is significantly related with high risk for injuries; nonetheless, which training variables truly associated with injuries is still inconclusive since there is no consensus of definition and systematic use of those definitions across studies (Nielsen et al., 2012). With regard to distance, Saragiotto et al in 2014 found in their systematic review of prospective studies that running more than 64 km per week is at the higher risk for injury. With regard to intensity, 3 to 7 times per week in male and 7 times per week in female is associated with risk of injury (Saragiotto et al., 2014). Harder running surfaces as well as wearing improper types of shoe or worn-out shoes have been implicated as risk factors for injuries but empirical evidence has not been conclusively established (Hreljac, 2005; Knobloch, Yoon, & Vogt, 2008; J. Taunton et al., 2003).

Anatomic variables or variations that are believed to contribute to injuries at the knee in runners comprise quadriceps angle (Q-angle), high or low longitudinal arch and subtalar pronation and limited hip, knee and ankle range of motion. Wen in 2007 provided nice summaries of anatomic risk factors retrieved from retrospective and prospective studies (D. Wen, 2007) whereas Saragiotto et al investigated only prospective studies in their most current systematic review (Saragiotto et al., 2014). The overall findings are as follows. Increased Q angle was significantly related with running related injuries. Specifically, large Q angle was associated with a development of patellofemoral pain (Lankhorst, Bierma-Zeinstra, & van Middelkoop, 2013). It should be noted that the consensus of a normal Q angle and methods of measuring Q angle has not been adopted. High and low longitudinal arch heights were associated with shin splints, compared with average arch heights. In general, high arch runner was related with lateral knee pain and bony injuries whereas low arch was associated with medial knee pain and soft tissue injuries. Greater subtalar pronation was associated with shin splints and iliotibial band syndrome.

Biomechanical variables that have been found to relate with running related injuries consist of weakness of hip muscle stabilization as well as kinetic and kinematic variables. Weakness of hip flexors, abductors (eg. gluteus medius muscle), the deep external rotators (piriformis, quadratus femoris, etc) were found in runners with patellofemoral pain (Ferber et al., 2009; D. Wen, 2007). With growing number of studies, kinetic and kinematic variables have been studied and related with running injuries include impact peak of vertical ground reaction force, instantaneous and average vertical load rate, knee flexion excursion, leg and joint stiffness and peak tibial shock. Greater instantaneous and average vertical load rate, sagittal knee stiffness as well as tibial shock was found in female runners with a history of tibial stress fractures, compared to control group (Milner et al., 2006; Milner, Hamill, & Davis, 2007; Zadpoor & Nikooyan, 2011).

In addition to the four categories of risk factors for related running injuries, one most recent systematic review included 11 prospective studies (Saragiotto et al., 2014) and 1 prospective observational research (Malisoux et al., 2014) found clear conclusion that a previous history of injury is the main risk factor.

Messier et al in 2008 and Malisoux et al in 2014 proposed a conceptual model of the etiologies of running related injuries or potential underlying mechanism of injuries. Even though different terminologies were used in their models, causes or risk factors for injuries are the interaction between *primary and complementary* factors. Figure 1.2 depicts a conceptual model of the causes of running injuries at knee joint which is modified from Messier and Malisoux's models (Malisoux et al., 2014; Messier et al., 2008). *Primary causes* include *training and biomechanical variables*. These variables are considered as primary because they have to be present or runners have to practice running for injury to occur. More importantly, they are preventable and modified with appropriate recommendation and intervention such as modification of training schedule, gait retraining strategies as well as a proper use of orthotic shoe insert, and footwear (O'Leary, Vorpahl, & Heiderscheit, 2008; Yeung & Yeung, 2001). *Complementary causes* consist of personal characteristics and anatomic variables which may be or may be not a cause of development of injury. Runners with those vulnerable variables have to be exposed to normal or extra load during running to manifest injuries. Unfortunately, all proposed risk factors and causes have been studied separately and in non-systematic way. Thus, no firm conclusion of actual underlying causes of overuse running related injuries can be made at this point.

1.3.3 Contributing and Risk Factors for Running Related Overuse Injuries at the Knee in Children and Adolescents without Neuromuscular Disorders

Only one research study reported the number of running related injuries in children and adolescents (Mehl, Nelson, & McKenzie, 2011). The data of running related injuries of children and adolescents 6 to 18 years old treated in Emergency departments were obtained through the National Electronic Injury Surveillance (NEISS) during 1994 -2007. Approximately 225,000 children and adolescent with 6 – 18 years of age were treated for running related injuries. Increasing in trend of running related injuries was 34% over the study period. Knee injury was the second most common sites of injury (15.3%). The most to least found types of injury were sprain and strain, fractures, soft tissues, lacerations, respectively. Onset and specific types of injuries were not studied and reported in this study. An estimated 75% of running related injuries occurred at school was found in children aged 6-14 years old. The common knee overuse injuries in pediatric and adolescent athletes who attended in organized and recreational sports consist of shin pain or medial tibial stress syndrome, Sinding-Larsen-Johansson (an apophysistis of lower pole of the patella), Osgood-Schlatter (a traction of the secondary ossification center of the tibial tubercle), patellofemoral pain and tibial stress fracture (Soprano & Fuchs, 2007).

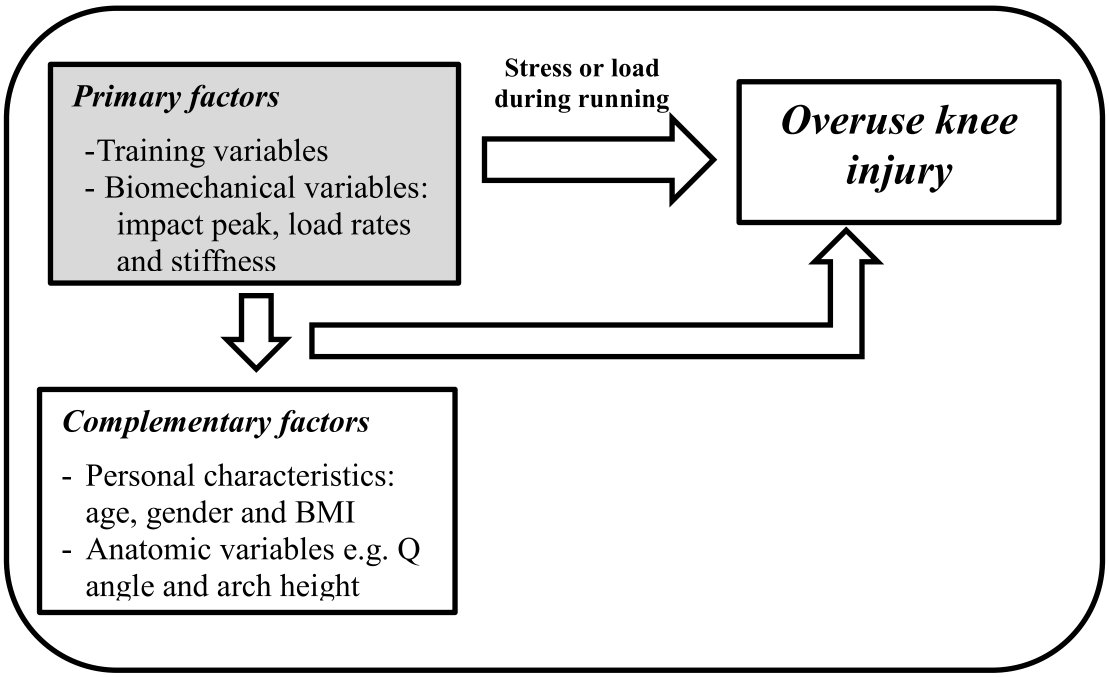

Figure 1.1 A conceptual model of risk factors for overuse knee injuries during running (adapted from Messier et al in 2008 and Malisoux et al in 2014)

Little research with respect to contributing and risk factors for overuse injury has been conducted in children. Based on anecdotal evidence, the risk factors contributing to overuse injury in children and adolescents, not specific only at lower extremity, in children are similar to those in adults. Only unique risk factors are *growth-related* factors. This growth related factors are very crucial in children because it may have detrimental consequences on normal growth and development. Two interconnected growth related factors have been considered as unique risk factors in children: 1) vulnerability of growth cartilage from repetitive stress and 2) growth spurt (DiFiori, 2010). Growth cartilage is present at articular cartilage, physes and apophyses (tendon-bone attachment sites). Growth cartilage is more prone to injury than mature bone (Blimkie et al., 1993; Flachsmann, Broom, Hardy, & Moltschaniwskyj, 2000). Repetitive shear and compression stress in conjunction with rapid changes during growth spurt may lead to overuse injury of growth cartilage. The National Athletic Trainer's Association addressed the importance of the injury surveillance, comprehensive physical examination and multidimensional approach for prevention of overuse injuries in pediatric population because approximately 50% of overuse injuries may be avoidable (McLeod et al., 2011).

1.3.4 Muscle Cocontraction

1.3.4.1 Operational Definition

Cocontraction or co-activation is simply defined as "*the simultaneous activity of various muscles acting around a joint"* (Kellis, Arampatzis, & Papadopoulos, 2003), p.229. Although this definition is commonly used to describe its role during movement, it does not specify how many or what muscles work to co-contract, especially the complex joints like shoulder or hip. Thus, quantification or clear explanation of cocontraction, including its role during movement is still underdetermined. Another definition that is easier to understand and study is *the simultaneous contraction of agonist and antagonist acting across the same joint and in the same plane* (Damiano, 1993; Falconer & Winter, 1985; Kellis, 1998). Based on this definition, an agonist or prime mover of the interested movement will be identified first and should across only one or two joints; its antagonist will be recognized and its force production should be in the exact opposite direction as the agonist; for example knee extensors is the agonist or prime mover during landing phase of running and knee flexor is the antagonist. Only two groups of muscles which locate or function oppositely can be simply studied for cocontraction.

1.3.4.2 Advantages Roles of Cocontraction during Movement

The actual roles of cocontraction in skilled movement are still controversial. However, three main roles may be concluded. First, muscle cocontraction may signify the achievement of a new motor skill. When learning a new motor skill, lots of unexpected joint movement may exist. An increased stabilization may be essential to reduce degree of freedom. To reduce degree of freedom, the joint becomes more stiffened which is controlled by increasing level of the antagonist muscle contraction or cocontraction. After a number of practices of a new motor skill, human body can predict those unexpected joint displacement which results in a reduced of stiffness or muscle cocontraction. Second, cocontraction plays special role when the accuracy and precision is critical for the motor task. One experiment found that when subjects were asked to perform finger movement with higher accuracy or precisely touched the endpoint, the degree of cocontraction was increased (as cited in (Damiano, 1993). Last, cocontraction supplements the mechanical properties of the ligaments in maintaining joint stability and protecting ligaments and joint injury during loading movement such as jumping and running (Fonseca, D.V, C.F., & Bricio, 2006). Noyes and Woo suggested that the normal load imposed to the knee joint during sport activities go beyond the tensile strength of anterior cruciate ligament (Noyes et al., 1989; Woo et al., 1991). Thus, another mechanism to protect joint injury should exist. Several researches suggested that this mechanism may be a control of cocontraction. The explanation is that cocontraction produces compressive forces which enhance contact between joint surface and increase joint stiffness. The modulation of joint stiffness via constant control of cocontraction is possibly the efficient mechanism to prevent joint injury during harmful loads (da Fonseca et al., 2004).

1.3.5 Excessive Knee Muscle Cocontraction in Children with CP during Walking and Running

Many researchers investigated muscle cocontraction in children with CP during walking but not yet during running. (Damiano, Martellotta, Sullivan, Granata, & Abel, 2000; Keefer et al., 2004; Unnithan, Dowling, Frost, & Oded, 1996; Unnithan, Dowling, Frost, Volpe Ayub, et al., 1996). Higher muscle cocontraction of knee and ankle muscles have been reported in children with spastic CP during walking when compared with children with TD, regardless of differences in gait protocols, sensor placement procedures, signal processing analysis of Electromyography (EMG), EMG normalization methods and cocontraction computational approaches. Higher level of muscle cocontraction during walking has been reported as excessive in children with CP when compared with children with TD. Excessive muscle cocontraction has many detrimental effects. Excessive cocontraction can increase joint stiffness and decrease agonist force production which restrains net joint movement. However, a link between muscle cocontraction and knee kinematic has never been investigated during either walking or running in children with CP. Since walking speed influences muscle activity in both children with TD and CP (Detrembleur, Willems, & Plaghki, 1997; Unnithan, Dowling, Frost, Volpe Ayub, et al., 1996), muscle cocontraction may be altered during running as well. There is evidence to speculate that muscle cocontraction may be high when children with CP run (Gross et al., 2013; Unnithan, Dowling, Frost, Volpe Ayub, et al., 1996). In Unnithan's study, cocontraction increased with increased walking speed (0.83 m/s vs. faster walking speed) was reported in both children with CP and TD (Unnithan, Dowling, Frost, Volpe Ayub, et al., 1996). In Gross's study, children with unilateral CP and children with TD were asked to walk at 3 different speed (slow, self-selected and fast speed), muscle cocontraction levels of three agonist-antagonist couples (rectus femoris/semitendiosus, vastus medialis/ semitendiosus and tibialis anterior/soleus) were increased when walking speed increased. They also found that levels of muscle cocontraction of involved-limbs were greater than those of non-involved limbs of children with unilateral CP and were more than limbs of children with TD, respectively.

1.3.6 Conceptual Model

A conceptual model was developed as a foundation for this research to guide the study of force attenuation during running in children with cerebral palsy (CP). Figure 1.2 depicts the conceptual model of this study which is called "*force attenuation mechanism*". This model is based on biomechanical perspective on risk factors contributing to injuries during running in individuals without neuromuscular disorder, which will be applied in children with CP. The risk factors that have been identified for running injuries can be placed into 3 categories, including structure or alignment of lower extremity, training errors and biomechanical factors. Abnormal static alignment is commonly seen in children with CP such as femoral and tibial torsion, knee abduction or foot overpronation (Morrell, Pearson, & Sauser, 2002). In addition to the training errors, biomechanical factors including impact force, load rate and stiffness are speculated as primary risk factors because they are high level of stress or external load that is directly applied to leg and body during running (Malisoux et al., 2014). If people do not run, high stress or external load does not occur. Therefore, training errors and biomechanical factors become the most interest areas of research because these factors can be preventable and modified by proper intervention (Cameron, 2010). Due to CP, interruption of neuromuscular control, such as abnormal muscle activation and excessive cocontraction, may be additional factors that place children with CP at high risk for injuries during running. Based on both aspects of knowledge, a conceptual model of "*impaired impact force attenuation*" in children with diplegic CP during running is proposed. Following is an explanation of the model and its components.

1.3.6.1 Impact Force and Load during Running

Impact force occurs when foot contacts the ground during walking or running. (Nigg, Cole, & Bruggemann, 1995). This force acts along bone from the floor and transmits from the lower extremity to the head. Typical impact force has positive effects on cartilage and bone growth. On the other hand, excessive impact force and load rates are speculated to be associated with musculoskelatal injuries such as stress fracture or early arthritis (Nigg et al., 1995). Ground reaction force (GRF) is commonly used to quantify the magnitude of impact force. Vertical GRF variables that have been studied and strongly correlate with various types of running related injuries include impact peak, load rate, and joint stiffness (Milner et al., 2006; Nigg, 2001; Nigg et al., 1995). In adults without neuromuscular disorders, normal values of impact peak and average load rate during running are approximately 1.2 to 3.5 body weights (BW) and 30-66 BW/seconds, respectively (Keller et al., 1996; Milner et al., 2006; Pohl, Mullineaux, Milner, Hamill, & Davis, 2008). Runners without neuromuscular disorders who do not have injuries, are able to attenuate high impact force to avoid injuries regardless of any environmental factors such as surfaces and shoe types. One of the ways to attenuate force is to alter kinematics, such as the knee flexion angle during ground contact (Derrick, 2004). Based on a recent running project in children with CP, children with CP demonstrated higher impact peak and load rate during running when compared with children with typical development (TD) although children with CP ran at lower speed (Orlin & Laibsirinon, 2010). Greater impact force in children with CP may be because the lack of force attenuation abilities that can be caused by impaired knee kinematic adaptation and abnormal neuromuscular control in some children with CP.

1.3.6.2 Impaired Knee Kinematic Adaptation to Impact Force

Running is a faster motion than walking during which the body is lifted higher. The leg contacts the ground faster and harder due to a quick drop of center of mass which leads to greater impact force. The knee is primary joint serving as force absorption during contact phase of running. Knee flexion is increased more during running than during walking. At initial contact (IC), knee is flexed in order to attenuate the higher impact force when the foot hits the ground. From loading response (LR) to midstance (MS), peak knee flexion is seen as a result of forward movement of tibia over foot. After midstance, knee flexion is gradually reduced before terminal stance (Novacheck, 1998) According to the study by Davids, et al, knee kinematic during running in children with diplegic CP was different in both peak angle and pattern. Children with CP contacted the ground with more flexed knee at IC and demonstrated less knee excursion of the entire contact phase, than children with TD (Davids et al., 1998). At IC, adolescents without neuromuscular disorder who run with larger knee flexion demonstrated lower impact force than running with typical amount of knee flexion. This method is called softer landing (McMahon et al., 1987). Based on the effective mass theory, a more extended knee contact angle can increase impact force and tibial shock (Derrick, 2004). The findings from the above two studies are inconsistent with the results found in Orlin's running project in children with CP (Orlin & Laibsirinon, 2010). Knee flexion at contact phase of running was increased throughout contact phase, but the impact force was still very high. A more fixed knee with less excursion and poor control of knee muscle suggest the inability to adapt and land softly to decrease impact force. This might be caused by poor neuromuscular control of children with CP.

1.3.6.3 Excessive Muscle Cocontraction in Children with CP during Running

Excessive muscle cocontraction or an excess antagonist muscle activity has been reported in both children and adults with Central Nervous System (CNS) damage (Ikeda, Abel, Granata, & Damiano, 1998; Lamontagne, Richard, & Malouin, 2000; Poon & Hui-Chan, 2009; Sgouros & Seri, 2002; Unnithan, Dowling, Frost, & Oded, 1996; Unnithan, Dowling, Frost, Volpe Ayub, & Bar-Or, 1996). Excessive muscle cocontraction during walking and running may have either positive or negative effect on children with spastic CP. First, it may accompany spasticity and muscle weakness during walking and running and to maintain postural stability or to prevent the stance leg from collapsing during stance phase while the opposite leg is in swing phase. A walking pattern of children with CP is similar to a pogo stick or a bouncing ball, instead of a typical pendulum pattern. Fonseca and co-workers proposed the term *dynamic resources* as a remaining system for walking in the absence of adequate muscle force for children with spastic hemiplegic. Although this pattern is not typical, it is functional. They stated that the leg of a child with spastic CP behaves like spring, which is characterized by the amount of stiffness of the integrated musculoskeletal system during the ground-contact phase. The stiffer system of legs of children with spastic CP may be attributable to excessive muscle cocontraction and stiffer soft tissue like muscles. A higher accumulation of collagen, that causes higher stiffness, has been reported in spastic muscles *(Booth, Cortina-Borja, & Theologis, 2001)).* Second, excessive muscle cocontraction may be detrimental because it can prevent normal joint movement resulting in too much joint stiffness. High joint stiffness has been purported to be a potential risk factor for running injuries (Butler, Crowell, & Davis, 2003). Too much stiffness highly correlates with injury, particularly with repetitive-high loading activity such as running (Butler et al., 2003). In children with spastic CP, muscle tightness, spasticity or excessive cocontraction can be considered as a cause of excessive stiffness of lower extremities. There is still unknown to what extent children with CP have excessive knee muscle cocontraction during running. Muscle cocontraction during running has never been examined in both children with TD and children with CP.

1.4 Preliminary Research

1.4.1 Inter-Reliability of Anthropometric Measures

The inter rater reliability of the measurement of anthropometric data used for joint center calculation in the video computerized gait analysis system was investigated to ensure that two examiners were able to perform the same anthropometric data measurement and obtain the similar data. Six anthropometric measurements as shown in Appendix A were measured by the two examiners. One novice physical therapist attended the study (tester 1). The other was a senior physical therapist with 20 years clinical experience (tester 2). Both of them were not trained how to use goniometer, tape measure and vernier caliper. The manual for anthropometric measurement was sent to them 2 days before data collection day. Testers reviewed and inquired regarding the manual.

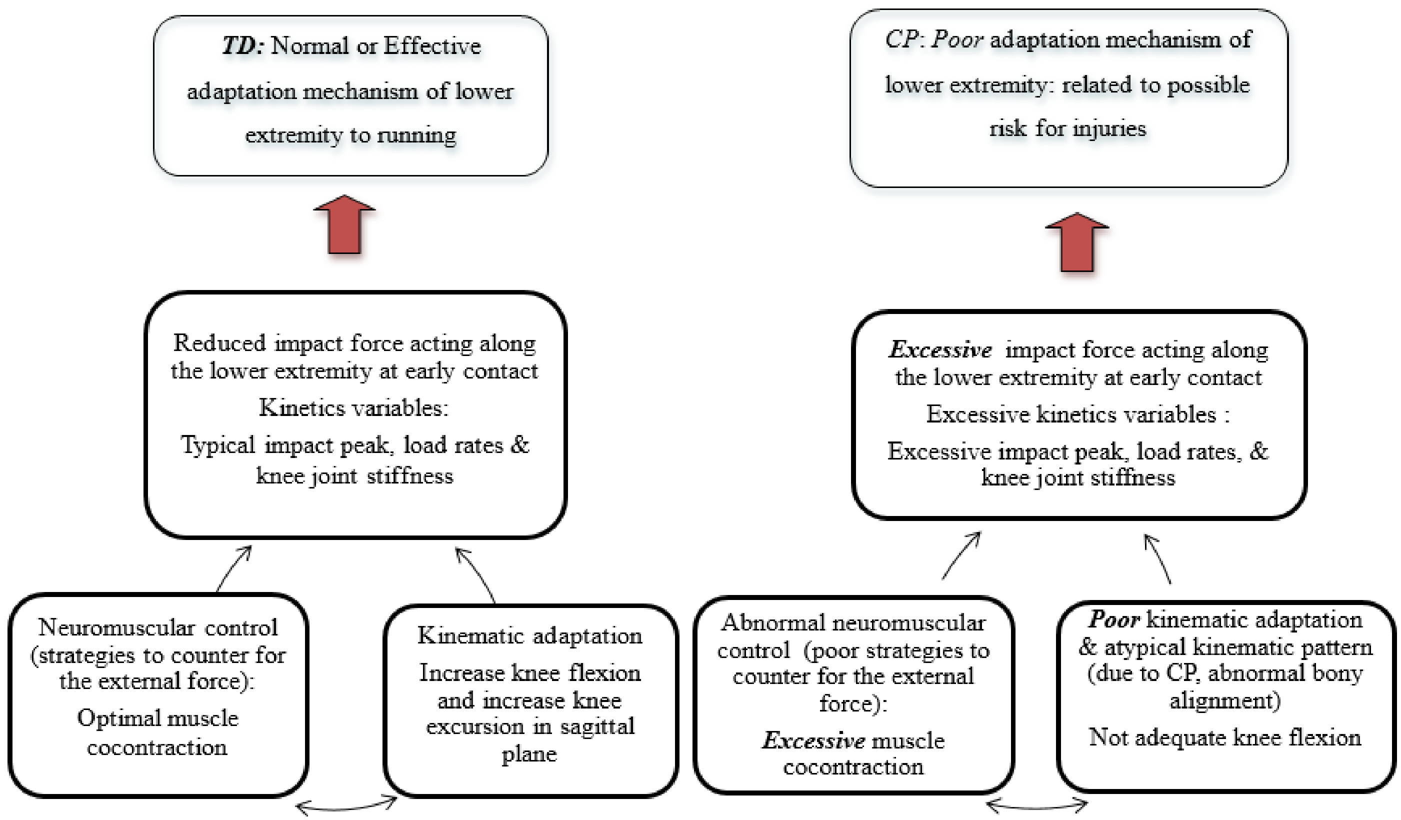

Figure 1.2 The conceptual framework of *impaired force attenuation mechanism* to reduce impact force during running in children with spastic cerebral palsy (CP)

Convenience sample of 10 participants was recruited into the study. Their mean age, height and weight were 27.8±8.6 years, 169.2±9.55 cm, and 67.1±23.5 kg, respectively. All of them were healthy. None of subjects was obese which was determined by Body Mass Index (BMI). Mean BMI was 22.5±3.4 kg/m^2. None had a history of musculoskeletal and neuromuscular disease. In addition, they all had normal strength and range of motion in the lower extremities and were willing to participate in the study. All participants were required to wear short. Both legs of all subjects were measured. Two testers performed six measurements on all subjects independently on the same day and were blinded from each other. One trial of all six measurements was orderly collected but it could start either from left or right leg.

Table 1.1 presents mean and standard deviation, Intraclass Correlation Coefficient ($ICC_{2,1}$) values and their 95% CIs, the corresponding error of measurement (SEM) and the minimal detectable change (MDC) of all seven measurements. The results suggest that the measurement methods of leg length assessed by using a tape measure and ankle width measured by using vernier caliper demonstrated good inter rater reliability and resulted in small SEM and MDC values. However, poor to moderate inter rater reliability was found for pelvic width, pelvic depth, knee width and tibial torsion. Values of SEM and MDC of all these measurements were consistent with their ICC. In order to improve inter rater reliability, palpation skills, practicing and training for performing measurements and revision of the anthropometric manual are needed.

Table 1.1 Mean, standard deviation (SD), standard error of measurement (SEM) and the minimal detectable change (MDC) of six anthropometric data, measured by tester 1 and tester 2.

Anthropometric data	**Tester 1 mean±SD**	**Tester 2 mean±SD**	**Main tester effect**	**$ICC_{2,1}$**	**95% CI of $ICC_{2,1}$**	**SEM**	**MDC_{95}**
Leg length (m)	0.88±0.06	0.89±0.06	F=0.48, p=0.51	0.98	0.91, 0.99	0.01	0
Pelvic width (cm)	23.77±1.39	25.38±1.12	F=54.40, p=0.00*	0.47	-0.07, 0.86	0.91	2.5
Pelvic depth (cm)	6.02±0.79	4.26±0.93	F=113.50, p=0.00*	0.27	-0.04, 0.72	0.73	2.0
Knee width (cm)	9.87±0.62	10.84±1.08	F=25.91, p=0.001*	0.49	-0.11, 0.85	0.61	1.7
Ankle width (cm)	6.31±0.52	6.42±0.50	F=1.11, p= 0.32	0.77	0.35, 0.94	0.24	0.7
Tibial torsion1 (degrees)	16.90±3.36	15.10±6.10	F=1.47, p=0.26	0.53	-0.50, 0.86	3.24	9.0
Tibial torsion2 (degrees)	15.95±3.31	15.2±6.03	F=0.35, p=0.57	0.68	0.12, 0.91	2.64	7.3

* = statistically significant at alpha level < 0.05

1.5 Research Plan

1.5.1 Research Design

Specific Aim 1: To compare kinetic variables related to running injures, as well as knee flexion angle and knee muscle cocontraction between children with diplegic Cerebral Palsy (CP) and children with typical development (TD)

Hypothesis 1-3: Independent variable is the clinical group (children with CP vs. children with TD)

Dependent variables: (1) impact peak, (2) average, and (3) instantaneous load rate, and (4) sagittal knee stiffness from initial contact to impact peak, (5) knee flexion angle at initial contact, (6) knee flexion angle at impact peak, (7) knee excursion from initial contact to impact peak, and (8) knee muscle cocontraction from initial contact to impact peak.

The research hypothesis is that children with CP will exhibit higher impact peak, load rate and sagittal knee stiffness at the phase of initial contact to impact peak, compared to children with TD. Children with CP will exhibit either greater or smaller knee flexion angle at initial contact and impact peak but smaller knee excursion from initial contact to impact peak, compared to children with TD. Children with CP will demonstrate higher knee muscle cocontraction from initial contact to impact peak, compared to children with TD

1.5.2 Participants

A convenience sample of children with diplegic cerebral palsy (CP) (GMFCS I and II) will be recruited from a school for special education. Children with typical development (TD) in Thailand will be invited to participate in this study. The range of age will be set at 7 to 15 years old for both groups. Children with diplegic CP will verbally report that they run at regular basis and also in their physical education classes. They will walk and run without any assistive devices. All children of both groups will be able to understand and follow all commands for this study. All children will provide verbal or written assent and guardians will provide written informed consent, as approved by the institutional ethics committees where the study will be conducted. Exclusion criteria for CP group include previous baclofen pump or dorsal rhizotomy surgery, lower extremity surgery, fractures, phenol or BOTOX injection within the past 6 months as well as previous history of hip subluxation or dislocation as documented in their medical record. A group of typical development will be healthy children who are to able and willing to run. They will be excluded if they have had lower extremity surgery, fractures within the past 6 months and a history of a musculoskeletal and neuromuscular disorder.

Sample size estimation for my study used the preliminary data from large running database (the "Run for Life") collected and granted approval by the primary investigator (Dr. Orlin, Margo). Only data from children with typical development (TD) and children with spastic diplgic cerebral palsy (CP) were obtained from the database and were utilized for sample size calculation. The selected variables that are key variables of my study include impact force, load rate, leg stiffness and knee flexion at initial contact and knee excursion from initial contact to the point of impact peak occurred. Muscle cocontraction data of children with TD and CP during running were not collected in Dr.Orlin's study and were not published anywhere else. For the priori power analyzes, power (1-β) was set at 0.8 and alpha was set at 0.01 (an initial alpha level of 0.05 with a Bonferroni correction factor of 7 dependent variables and 2 involved legs of each group:0.05/28). The Cohen *d* was used to calculate the effect size indices for mean analyzes between 2 groups of all dependent variables. For all 3 aims, impact peak and joint stiffness required about 30 subjects for each group. Knee flexion angle and excursion required below 10 subjects.

Ten subject with each group of children TD and spastic diplegic CP will be investigated in the proposed study. This number is practical and realistic, considered a length of time to complete my dissertation.

1.5.3 Instrumentation, Tests and Measures

Knee joint kinematic during running will be collected by using an eight-camera VICON MX-T series gait analysis system (Oxford Metrics, Oxford, UK) through capture of reflective markers attached on specific anatomical landmarks according to the lower body of plug-in-gait model version 1.9 (VICON: Plug-in Gait-Foundation Notes). This system will record the three-dimensional position data or kinematics. The computerized video-camera gait analysis system has been extensively used in the assessment of gait deviation and in evaluating the effect of interventions in children with spastic CP (Gage, 1993) because it is more accurate than visual assessment (Mackey, Lobb, Walt, & Stott, 2003) and has high intra-subject reliability (Steinwender et al., 2000). Moderate to high intra-subject repeatability (coefficient of multiple correlation: CMC) of both within day and between day of all joint range of motion during gait was reported in children with spastic diplegia. Particularly, CMC values of knee joint angles in sagittal plane were above 0.95 (Steinwender et al., 2000).

Vertical ground reaction force will be collected by using three AMTI force plates model OR6-7 2000 (Width 18.25 x Length 20 x Height 3.25 in) embedded in the middle of a 15 m walkway. The force plate consists of a six-channel strain gage amplifier system. An amplifier gain set at 1000 Hz and a second-order critically damped low-pass filter with a cut-off frequency of 10 Hz. The bridge excitation is set at 10.00 volts DC to all 6 channels. Prior to data collection, the bridge will be balanced for each channel and set at zero by the main control center and ensure time-synchronization with VICON system. Impact peak, average load rate, instantaneous load rate will be subsequently calculated from vertical GRF by using custom written LabView software (National Instruments Corporation, Austin, TX, USA).

Knee muscle cocontraction will be collected by using a Zero wire surface electromyography (EMG) (Aurion, Inc., Milano, Italy) and surface electromyography electrodes. A Zerowire is a wireless telemetry system (amplifier input impedance: 20MOhm; frequency response: 10-1000 Hz, common mode rejection ratio: 90 bB) that is time synchronized with the VICON MX system. The EMG is hardware filtered using an analog RC filter from 10 to 500 Hz. The types of circular surface electrodes will be bipolar sensor and pre-gelled Silver/silver chloride with a conductive area of 10 mm^2 (Kendall Meditrace; Tyco Healthcare, Hamshire, UK)

1.5.3.1 Anthropometric Measures

Anthropometric measurements which include weight, height, leg length, knee width and ankle width will be performed by using a lab vernier caliper based on standardized protocol for Plug-in Gait model. Hamstring muscle length will be measured by popliteal angle test. Gastroc-soleus muscle length will be collected as a degree of ankle dorsiflexion while a knee is fully extended. Tibial torsion will be measured. All participants will receive passive range of motion of hip, knee and ankle joints to determine joint integrity.

1.5.3.2 Data Processing and Reduction

After data collection, VICON NEXUS software, version 1.8.4 will be used to post-process in order to generate temporal-spatial data, marker and kinetic data. Initial contact phase and toe-off of running gait events will be automatically detected by using a presence and absence of vertical GRF (set at 10 N). Later, knee flexion angle at initial contact and knee flexion excursion from initial contact to impact peak will be extracted manually into an excel spreadsheet. Average knee joint stiffness from initial contact to impact peak will be computed at the change in joint moment divided by the change in joint angle (Farley & Gonzalez, 1996). Gait Deviation Index (GDI) will be used to determine more and less involved leg of children with CP (Schwartz & Rozumalski, 2008). Nine kinematic angles of pelvic and hip angles in all 3 planes, knee flexion/extension and ankle dorsiflexion/plantarflexion and foot progression during walking will be used to calculate Gait Deviation Index (GDI). The GDI measures a distance away from the average control gait. GDI scores ≥ 100 indicate a subject whose gait is close to the control average. Every ten points falls below 100 GDI equals one standard deviation away from the control mean. In other words, lower GDI indicates more involved leg in children with Cerebral Palsy.

Vertical impact peak, time to impact peak, average and instantaneous load rates will be calculated and normalized to body weight (BW) after kinetic data will be filtered at 50 Hz using a fourth order low-pass Butterworth filter by using custom written LabView software (National Instruments Corporation, Austin, TX, USA) program as courtesy by the gait lab at University of Delaware . If there will be no distinct impact peak in the vertical GRF of forefoot strike runner, vertical impact peak will be identified corresponding to the mean time when the impact peak occurred in the rear foot strike pattern of children with TD in the large running database and from this study (Boyer, Rooney, & Derrick, 2014). Average and instantaneous load rates will be calculated between 20 and 80% of the period between initial contact and impact peak (Milner et al., 2006).

The raw EMG signal for each muscle will be full wave rectified and low pass filtered at 4 Hz (digital 2^{nd} order Butterworth at 20 Hz cut-off frequency) to create a linear envelope for cocontraction index analysis using another custom written LabView software (National Instruments Corporation, Austin, TX, USA). Knee muscle cocontraction will be extracted, corresponding to the mean time from initial contact to when impact peak occurred in vertical GRF. Numerous methods of EMG data processing have been used in order to quantify muscle cocontraction during gait in children with CP (Damiano et al., 2000; Ikeda et al., 1998; Keefer et al., 2004; Unnithan, Dowling, Frost, & Oded, 1996). An equation of Cocontraction Index (CCI) derived by Falconer and Winter will be used in this study (Falconer & Winter, 1985). This approach is chosen because of 2 reasons; 1) it is aligned with the operation definition in this study; 2) to date, it is the only one approach that is used to investigate knee cocontraction in children with CP during walking, similarly to this study (Keefer et al., 2004).

$$\text{Cocontraction Index (CCI)} = \frac{2 \text{ x common area between RF and BF}}{\text{Area of RF + area of BF}}$$

In this equation, the linear envelop area of RF and BF are areas under the curve of individual EMG activity of Rectus Femoris and Biceps Femoris. The Common area between RF and BF is the area under the curve when RF and BF area is overlaid (the RF and BF both active concomitantly). The common area is multiplied by 2 because it accounts for counter balance moment of the agonist and antagonist or the extra effort the agonist muscle has to exert in order to overcome the antagonist activity. Cocontraction index (ICC) is always less than 1 or 100%.

1.5.4 Procedure

Prior to data collection, all guardians will provide written informed consent and children will provide either verbal or written assent based on their ability to write their names. The procedure and risk during data collection will be explained to the guardian and child. Demographic information will be obtained from their medical records at school with guardian's written permission prior to and verbally obtained on collecting date.

The surface EMG electrode's location, placement, and fixation techniques will be followed a guideline from the Surface Electromyography for the Non-Invasive Assessment of Muscles (SENIUM) European recommendations for surface electromyography (Hermens et al., 1999). Before the electrodes will be placed, area of electrode placement will be cleaned with alcohol pad. The surface EMG electrodes will be placed over the muscle bellies of rectus femoris and biceps femoris bilaterally and held securely with Coban Co-flex. The inter-electrode distance will be within 1 cm.

Fifteen reflective markers will be placed on anatomical and segmental landmarks of both legs as specified by Plug-in Gait lower body model version 1.9 (VICON: Plug-in Gait-Foundation Notes) which include the midpoint of the sacrum, anterior superior iliac spine, lateral side of mid thigh, lateral femoral epicondyle, lateral side of mid shank, lateral malleolus, head of second metatarsal head and mid posterior calcaneous. All markers will be held securely on the skin with hypoallergenic tape.

Children will be asked to walk barefoot along 15 meter walkway in order to use for Gait Deviation Index (GDI) calculation. All participants will be requested to run barefoot at preferred jog speed. At least 3 trials of running barefoot passing the middle of three force plates will be collected at 100 Hz by using a VICON MX-T motion analysis capture system. Ground reaction force (GRF) data and EMG data will be sampled at 1000 Hz and time-synchronized with a VICON MX motion analysis capture system. Children will have as many rest breaks as they prefer during data collection.

1.5.5 Data Analysis

Average and standard deviations of age, weight, height, body mass index (BMI) will be used to describe subject characteristics. Three trials of running velocity will be averaged. Normality of data will be determined by checking the skewness of the data and also used a test of normality.

Independent t- test or Mann-Whitney U test will be used to compare dependent variables between clinical groups (CP vs. TD). Alpha is set at 0.05. Either left or right legs of children with TD will be randomly selected to compare with less and more involved legs of children with CP.

1.6 Potential Problems and Alternative Strategies

Correct foot placement on the force platform without altering gait pattern is very difficult in individuals without neuromuscular disorders, even much more difficult in children with CP based on personal experience as research assistance in a large preliminary study (52 participants). This issue can cause exhaustion and fatigue in children. Data collection can be lengthy. Children will take a rest break as often as they prefer. Participants with CP will attend school and only available 2 hours a time. Therefore, they will re-participate with a new consent form if the data collection will not be completed within 2 hours. Children may experience mild discomfort from hypo allergic tape, gel and Coban Co-flex that will be used to secure markers and surface EMG electrode. Afterward, skin will be thoroughly clean with alcohol pad. If they will report allergic reaction with alcohol, water and soap will be used alternatively. Skin check for redness and/or irritation will be done before children will leave the lab. Children may experience exhaustion, muscle fatigue or discomfort/pain at the foot or leg after running barefoot. A researcher will inquire children at the end of data collection and also check with a school nurse or physical therapist at the end of day to ensure all aforementioned symptoms will be completely resolved. Children in both groups may fall during testing. Another research assistant will follow children concomitantly while running to ensure their safety.

1.7 Resources

1. The Motion Analysis Lab is equipped with at least 6 video cameras that are time synchronized with a minimum of two force plates and 4 channels of electromyography device.
2. A research assistant assists during data collection when participants will run to ensure safety if they will fall or lose balance.
3. The materials for data collection consist of electromyography (EMG) electrodes, Coban Co-flexes, hypoallergenic tapes, alcohol pads, tank tops and shorts
4. The software packages for data post-processing include the LabView software (National Instruments Corporation, Austin, TX, USA) and the MatLab R2008a software (The Mathworks, Natick, MA, USA).

1.8 Timeline

This research will be finished within 1 year. The plan is below:

	Month 1	Month 2	Month 3	Month 4	Month 5	Month 6	Month 7	Month 8	Month 9	Month 10	Month 11	Month 12
1.Review literature and prepare for all materials												
2.Recruit subjects												
3. Collect data												
4. Process and analyze data												
5.Write report and manuscript												

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CHAPTER 2

Impact Force, Knee Muscle Cocontraction, Knee Stiffness and Knee Angle during Early Contact of Running in Children with Diplegic Cerebral Palsy

Impact Force, Knee Muscle Cocontraction, and Knee Angle during Early Contact of Running in Children with Diplegic Cerebral Palsy

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2.1 Abstract

Purpose: Some children with diplegic cerebral palsy (CP) are able to run. The inability to effectively attenuate impact force during running may predispose them to lower extremity injuries. The purpose of this study was to compare impact peak, load rates, knee stiffness, and knee contact angle during early stance phase of running between children with CP and children with typical development (TD).

Methods: Six children with CP and eight children with TD, aged 7 to 15 years, ran at a comfortable speed in the gait laboratory equipped with an eight-camera motion analysis system, three force plates and a wireless electromyography. Variables of interest were impact peak, load rates, knee stiffness, and knee contact angle which were calculated from a period of initial contact to the impact peak of vertical ground reaction force.

Results: Impact force (~1.9 BW), average and instantaneous load rates (~110-168 BW/s) were significantly higher in children with CP, compared to those with TD (~0.9 BW and ~60 BW/s, respectively) despite similar running speeds. No other variables were different between the two groups.

Conclusion: High impact force and load rates may be used to identify children with CP who may be at risk for lower extremity injuries. Parents and individuals working with children with CP should consider excessive impact force and load rates if children complain of any pain during and after running. Proper footwear, orthoses, and running retraining may be intervention options to reduce impact forces and decrease the risk for injuries.

Keywords: Cerebral Palsy, Running, Impact peak and Knee contact angles

2.2 Introduction

Running is a simple form of exercise that is possible for many children with ambulatory diplegic CP (Brunton & Bartlett, 2010; Maher, Williams, Olds, & Lane, 2007; Palisano et al., 1997). However, due to concerns about injuries, running is not often encouraged for children with CP. The primary concern for running with CP is the potential long-term damage to lower extremity joints. Repetitive high impact force during running may be a predisposing factor to the development of pain and premature musculoskeletal damage, accelerating a decline in other ambulatory functional activities such as walking. Several factors contribute to concerns about developing lower extremity injuries in children with CP. Children with CP commonly present with atypical leg-foot alignments such as excessive femoral and tibial torsion, knee abduction or foot over-pronation (Driscoll & Skinner, 2008; Morrell, Pearson, & Sauser, 2002) and abnormal leg-foot alignment has been reported as one of the causes of running related injuries in runners (Saragiotto et al., 2014; Wen, 2007). Other variables strongly correlated with running related injuries include atypically high vertical impact peak and load rate, and excessive leg and joint stiffness (Hreljac, 2005; Milner, Ferber, Pollard, Hamill, & Davis, 2006). Runners who are unable to successfully attenuate the increased impact force and load rate have been shown to experience injuries (Grimston, Engsberg, Kloiber, & Hanley, 1991; Grimston, Nigg, & Fisher, 1993; Hreljac, 2005; Milner et al., 2006). Orlin and Laibsirinon found higher impact peaks and load rates during the stance phase of running in children with CP, compared to children with TD (Orlin & Laibsirinon, 2010). Therefore a combination of abnormal leg-foot alignment and high impact force, both likely to be present in children with CP, may place those runners at higher risk for injuries.

In individuals without neuromuscular disorders, increased knee flexion angle at early contact phase of running has been suggested as a strategy to attenuate impact force and consequently reduce injury potential (Derrick, 2004; Hreljac, 2004, 2005). In children with CP, the high impact force and load rates may be a result of an inability to attenuate forces because of an inability to modify knee flexion angle at early contact. One study has reported a higher knee flexion angle at initial contact but smaller knee excursion throughout contact phase of running in children with CP, compared with children with TD (Davids, Bagley, & Bryan, 1998). A higher knee flexion angle at initial contact should decrease the impact force but instead, children with CP demonstrated higher impact force and load rates (Orlin & Laibsirinon, 2010). This seeming contradiction may indicate a stiffer knee joint in children with CP, often attributed to excessive knee muscle cocontraction.

Excessive cocontraction of knee muscles has been investigated and reported in children with diplegic CP during walking (Keefer et al., 2004; Maltais et al., 2004; Unnithan, Dowling, Frost, Volpe Ayub, & Bar-Or, 1996) but has yet to be investigated during running. Excessive muscle cocontraction causes a stiff knee joint, which restricts movement and may contribute to the inability to modify knee contact angle, resulting in ineffective absorption and attenuation of increased impact force and load rates during running. However, this is only a speculation since no one has yet systematically investigated the risk factors for running injury (impact force, load rate, knee joint stiffness, knee flexion angle, and knee muscle cocontraction) in children with CP, particularly those known to be common for such children. This reflects a critical gap in our understanding about the risk factors related to running for children with CP. Lack of this knowledge can lead to unrealistic concerns about the development of injuries due to running in children with CP and may limit health professionals from identifying true risk factors for musculoskeletal injuries that may be sustained during running.

Thus, this study will begin to investigate kinetics including impact peak, load rates, knee stiffness, and knee flexion angle during early contact phase of running in children with diplegic CP. In the long term, knowledge of kinetics, knee flexion angle and knee muscle cocontraction during running can be used to inform clinical decision making, identifying risk and encouraging safe participation in running for children with CP. The ***purpose of this study*** was to compare kinetics, knee flexion angle and knee muscle cocontraction between children with diplegic CP and children with TD at a period of initial contact (IC) to impact peak (IP). The ***central hypothesis*** was that compared to children with TD, children with diplegic CP will demonstrate higher impact peak, higher load rates, and higher knee joint stiffness during IC to IP. Children with CP will demonstrate higher knee muscle cocontraction. Children with CP will also display different knee flexion angle at IC and at IP, and smaller knee excursion from IC to IP.

2.3 Materials and Methods

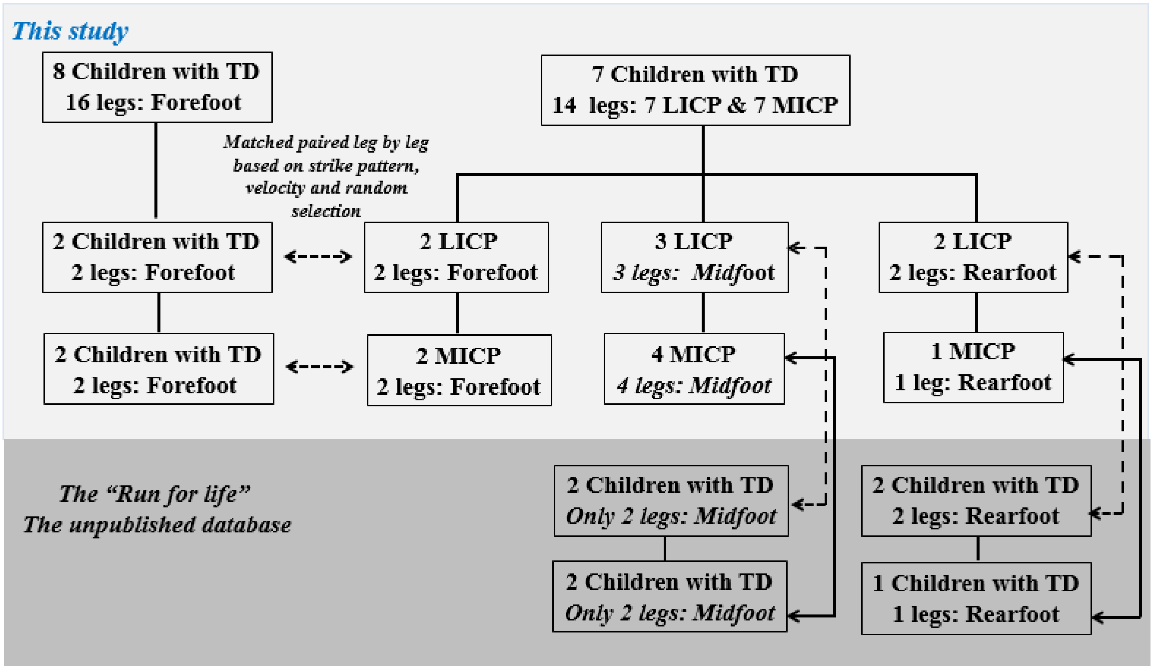
Participants

Seven children with diplegic CP classified at level I or II on the Gross Motor Function Classification System (GMFCS) were recruited from a school for special education in Pathum Thani province, Thailand. They were excluded if they had previous baclofen pump or dorsal rhizotomy surgery, lower extremity surgery, fractures, phenol or BOTOX injection within the past 6 months, or previous history of hip subluxation or dislocation. All children with CP were able to walk and run independently without assistive devices. All children with diplegic CP were interviewed about their running activities and were observed during physical education classes. All children with CP ran on a regular basis such as during gym class, recess, or play. Physical examination of muscle length, muscle strength and static lower extremity alignment were performed in all participants. Hamstring tightness was not found as determined by ≥125 degrees of popliteal angle (Kuo, 1997), but some gastronemius-soleus tightness (as determined by -10 to 20 degrees of dorsiflexion with full knee extension (Keenan et al., 2004) was identified in all children with CP. Eight children with TD also participated as the comparison group for this study. All children provided verbal or written assent and guardians provided written informed consent. This study was approved by the ethics committee of Thammasat University and Sirindhorn National Medical Rehabilitation Center (SNMRC), Thailand. For purposes of comparison, the more and less involved legs of children with CP were identified by using the gait deviation index (GDI) (Schwartz & Rozumalski, 2008) and compared leg by leg with those of children with TD. The GDI process is further described in the data analysis section below.

Kinetics and knee flexion angles of four de-identified children with typical development from the unpublished database (the primary investigator granted the access) were included in this study because of differences in strike patterns (rearfoot, midfoot and forefoot) that were used by the children with CP. Different foot strike patterns have been shown to alter impact peak, load rate and joint stiffness, which could confound the results of this study (Almeida, Davis, & Lopes, 2015). This unpublished database (called “Run for Life”) was constructed at Philadelphia Shriners Hospital during 2006-2010 with data collected by Dr. Margo Orlin with Ms. Laibsirinon serving as a research assistant. The same procedures and similar VICON motion analysis systems were used between the current study and the “Run for Life” study.

For the more involved leg of children with CP (MICP), one out of the seven legs contacted the ground with the rearfoot, four with midfoot and two with forefoot. For the less involved leg of children with CP (LICP), two out of the seven legs contacted the ground with the rearfoot, two with midfoot and three with forefoot. However, all sixteen legs of the eight children with TD from this study struck the ground with the same pattern, which was forefoot. Therefore, the midfoot and rearfoot striking legs of the children with CP were matched paired one by one with those same striking patterns for children with TD from the “Run for Life” study. If both legs of a child with TD had the same strike pattern, only one leg was randomly selected to compare with the leg of a child with CP. In total, six LICP and 5 MICP, compared to those legs of children with TD were available because of limited number of children with TD who ran with midfoot pattern. Figure 1 presents a flowchart of this matching protocol.

Demographics including age, weight, height and body mass index (BMI), and running velocity of children with CP and children with TD (from this study and the Run for Life study) are presented in Table 2.1. No statistically significant differences of age, weight, height, BMI and running velocity were found between the two groups.



*Dr.Orlin' s the "Run for life" study. This author was granted an access to this database

Figure 2.1 Flowchart of matched paired leg by leg between children with CP from this study and children with TD from the "Run for Life" unpublished database

Table 2.1 Demographic of participants and running velocity

Participant demographic & average running velocity	TD≠					CP				
	Age (years)	**Height (m)**	**Weight (kg)**	**BMI (kg/m^2)**	**Running velocity (m/s)**	**Age (years)**	**Height (m)**	**Weight (kg)**	**BMI (kg/m^2)**	**Running velocity (m/s)**
	9	1.35	28.9	15.9	1.9	11	1.18	25.0	18.0	1.3
	7	1.33	30.9	17.5	2.6	12	1.47	34.0	15.7	2.1
	9	1.44	28.8	13.9	3.0	10	1.31	20.9	12.2	1.7
	10	1.40	45.6	23.3	3.0	13	1.04	38.4	35.5	2.2
	9	1.37	29.7	15.8	2.1	11	1.44	44.4	21.4	2.0
	12	1.60	76.2	29.8	2.7	9	1.29	25.00	15.0	1.7
	12	1.47	53.8	24.9	1.8					
	12	1.45	37.1	17.6	2.1					
Mean	**10.0**	**1.4**	**41.4**	**19.8**	**2.4**	**11.0**	**1.3**	**31.3**	**19.6**	**1.8**
SD	**1.9**	**0.1**	**16.7**	**5.5**	**0.5**	**1.4**	**0.2**	**9.1**	**8.4**	**0.3**

≠ 2 children with TD was included from this study and 6 children with TD was included from the unpublished database

Instruments

Three instruments were used for data collection: 1) an eight video-camera VICON MX-T series gait analysis system with passive markers, collecting at 100 Hz (Oxford Metrics, Oxford, UK); 2) three AMTI force plates model OR6-7 embedded in the middle of a fifteen-meter walkway, collecting at 1000 Hz (AMTI Inc, MA, USA). The force plate consists of a six-channel strain gage amplifier system. The bridge excitation was set at 10.00 volts DC to all 6 channels. The bridge was balanced for each channel and set at zero by the main control center and 3) a Zero wire surface electromyography (EMG) (Aurion, Inc., Milano, Italy) along with a 10 mm^2 conductive area of bipolar and pre-gelled silver/silver chloride surface EMG electrodes (Kendall Meditrace; Tyco Healthcare, Hamshire, UK). Prior to data collection, all instruments were calibrated and time synchronized.

Procedures

Prior to data collection, all children changed into black tank tops and black shorts that were provided by the researcher. Fifteen reflective markers were placed on anatomical and segmental landmarks of both legs as specified by Plug-in Gait lower body model version 1.9 (VICON: Plug-in Gait-Foundation Notes). All markers were held securely on the skin with hypoallergenic tape. The surface EMG electrode's location, placement, fixation and normalization techniques were followed a guideline from the Surface Electromyography for the Non-Invasive Assessment of Muscles (SENIUM) European recommendations for surface electromyography (Hermens et al., 1999). The areas of electrode placement were cleaned with alcohol prior to placement. The surface EMG electrodes were placed over the muscle bellies of rectus femoris (RF) and biceps femoris (BF) bilaterally and held securely with Coban Co-flex. The inter-electrode distance was within 50 mm to prevent crosstalk (Hermens, Freriks, Disselhorst-Klug, & Rau, 2000). In order to establish a reference for EMG data, maximal voluntary contraction (MVC) of quadriceps and hamstrings of both legs of all children were collected by having children perform three isometric contraction with a three-minute resting between each trial. Average of three trials of MVC were used for normalization before the calculation of cocontraction index (CCI).

After standing calibration trials were collected in all children, they were asked to walk barefoot along a fifteen-meter walkway. Then, all participants were requested to run barefoot at their preferred speed. At least three acceptable running trials, in which each leg contacted the middle of force plates were collected. Children were allowed as many rest breaks as they required during data collection.

Data analysis

After data collection, more and less involved leg of children with CP (MICP and LICP, respectively) were identified by using the GDI (Schwartz & Rozumalski, 2008) calculated from kinematic data during walking trials. The GDI of each leg of the children with CP was computed by comparing nine kinematic measures against the reference data from children with TD in this study. A GDI $\geq$ 100 indicated non-pathological gait and each 10-point below 100 indicated 1 standard deviation from control reference data. Lower

GDI scores (≤ 100) denoted greater deviation during gait from the control reference data. Therefore, lower GDI indicated the more involved leg. Mean and standard deviation (SD) for GDI was 80.9±4.1 for less involved legs of CP and 77.8±3.48 for more involved legs of CP.

Kinetic data including impact peak and load rates were filtered at 50 Hz using a fourth order low-pass Butterworth filter by using custom written LabView software (National Instruments Corporation, Austin, TX, USA), as courtesy by the gait laboratory at University of Delaware. Vertical impact peak, average and instantaneous load rates were calculated and normalized to body weight (BW). The *impact peak* was the maximum amplitude of vertical ground reaction force during the impact phase (Nigg, Cole, & Bruggemann, 1995). All children with TD in this study ran with forefoot strike as detecting by using Foot Strike Angle (SFA) (Altman & Davis, 2012; Pontzer et al., 2014). Because no distinct vertical impact peak was identified in the children with TD striking the ground with forefoot, the *vertical impact peak* was defined as corresponding to the mean time when impact peak occurred in the children with TD striking the ground with rearfoot based upon the Run for Life database. The timing of the impact peak of vertical GRF was used to determine the period from IC to IP. *Average loading rates* were assessed from slope between 20 and 80% of vertical GRF from the period from IC to IP or the first change of the slope of vertical GRF (Milner et al., 2006). *Instantaneous loading rate* was determined as the peak change in force over the same time interval for which average loading rate was identified.

VICON NEXUS software, version 1.8.4 was used to post-process marker trajectories and GRF data in order to generate knee flexion angles and moments. Marker trajectories and GRF were low pass filtered at 8 Hz using a fourth order Butterworth filter. Cut-off frequency of 8 Hz was chosen by using a residual analysis of XYZ directions of all markers while retaining 95% of the signal (comparing residuals of the differences between filtered and unfiltered signals at several cut-off frequencies) (Winter, 2009). Knee flexion angle at IC and at IP, and knee excursion from IC to IP were extracted manually into an excel spreadsheet. *Average knee joint stiffness* was computed as the change in net knee joint moment divided by the change in knee joint angle from IC to IP (Farley & Gonzalez, 1996)

Even though CCI was one of variables of interest and was part of the original study plan, the unpublished database did not include EMG data used for CCI calculation. Knee muscle cocontraction from only four legs of two children with CP and four legs of two children with TD (who ran with forefoot) from this current study were available. Therefore, the CCI was not statically analyzed and not presented in the result and discussion sections.

Statistical analysis

Two sets of comparison of each variable of interest were statistically analyzed 1) less involved leg of CP (LICP), compared with children with TD, and 2) more involved legs of CP (MICP), compared with children with TD. Mann-Whitney U test, with significance level set at 0.05 was used to compare demographic data and variables of interest between children with CP and children with TD. Approximate effect sizes of r ($r = z/\sqrt{N}$) was calculated (Filed, 2013). The probability that a randomly selected subjects from the CP group would have higher magnitude of dependent variables than a randomly selected TD subjects was also computed and reported.

2.4 Results

Table 2.2 presents two comparisons for the variables of interest: 1) those with TD compared to the less involved leg of those with CP (LICP) and 2) those with TD compared to the more involved leg of those with CP (MICP).

Three variables compared between LICP and children with TD were significantly different. Impact peak and load rates were higher in the children with CP. One variable, impact peak, compared between MICP and TD was significantly different, again higher in the children with CP. Large effect sizes between 0.6 and 0.8 and probabilities between 86% and 97% that a randomly selected subject from the CP group would be higher than a randomly selected TD subject were found for all four significantly different variables. Median values of impact peak of children with CP were approximately twice as high as those of children with TD regardless of leg involvement. Although in MICP neither load rate was significantly different from children with TD, the median values of both load rates were twice as high as those of children with TD. Small to medium effect sizes and greater than 60% probability for both load rates between children with MICP and TD were found. None of the other variables were significantly different.

Table 2.2 Kinetics during early contact phase of running in children with TD and children with CP

Variables Median (min, max)	TD values* (2R, 2M, 2F) †	CP: Less involved leg (2R, 2M, 2F) †	p-value***	Effect size# between TD and CP less involved leg (probability)##	TD values** (1R, 2M, 2F) †	CP: More involved leg (1R, 2M, 2F) †	p-value***	Effect size# between TD and CP More involved leg (probability)##
Impact peak of vertical GRF (BW)	0.9 (0.5, 1.3)	1.9 (1.1, 2.5)	***0.004***	0.8 (0.97)	0.8 (0.8, 2.1)	1.9 (1.4, 3.1)	***0.05***	0.6 (0.88)
Instantaneous load rate of vertical GRF (BW/s)	62.3 (17.3, 131.4)	167 (77.4, 247.8)	***0.04***	0.6 (0.86)	68.2 (28.7, 267.3)	129.9 (62.2, 244.2)	0.47	0.2 (0.64)
Average load rate of vertical GRF (BW/s)	49.9 (10.3, 116.3)	167.8 (54.3, 224.7)	***0.04***	0.6 (0.86)	44.3 (19.3, 241.2)	109.6 (47.2, 199.4)	0.35	0.3 (0.68)
Knee Joint stiffness (Nm/degrees)	0.05 (0.03, 0.10)	0.07 (0.05, 0.15)	0.17	0.4 (0.72)	0.06 (0.03, 0.07)	0.05 (0.03, 0.07)	0.46	0.2 (0.32)

* values from children with typical development that were matched pair one by one with ***less*** involved legs of children with CP

** values from children with typical development that were matched pair one by one with ***more*** involved legs of children with CP

*** Mann-Whitney U test was used to compare all variables between children with CP and TD

† Number of legs with different strike patterns. R is Rearfoot, M is Midfoot and F is Forefoot

r effect size (r = $z/\sqrt{N}$) and ## The probability that a randomly selected subjects from the CP group would be higher than a randomly selected TD subjects

Table 2.3 Knee angles during early contact phase of running in children with TD and children with CP

Variables	TD values* (2R, 2M, 2F) †	CP: Less involved leg (2R, 2M, 2F) †	p-value***	Effect size# between TD and CP less involved leg (probability)##	TD values** (1R, 2M, 2F) †	CP: More involved leg (1R, 2M, 2F) †	p-value***	Effect size# between TD and CP More involved leg (probability)##
Knee flexion angle at Initial Contact (IC) (Degrees)	22.7 (6.7, 29.1)	19.1 (2.9, 39.1)	0.52	0.2 (0.39)	18.6 (9.1, 21.6)	14.7 (6.8, 33.6)	0.60	0.2 (0.40)
Knee flexion angle at Impact Peak (IP) (Degrees)	25.4 (15.6, 30.8)	24.28 (4.4, 40.8)	1.0	0.0 (0.50)	21.1 (19.0, 27.5)	19.1 (8.4, 35.8)	0.47	0.2 (0.36)
Knee range from IC to IP (Degrees)	3.1 (1.8, 10.5)	4.7 (1.6, 9.5)	0.87	0.1 (0.47)	4.9 (1.2, 10.2)	3.0 (2.6, 5.2)	0.75	0.1 (0.44)

* values from children with typical development that were matched pair one by one with ***less*** involved legs of children with CP

** values from children with typical development that were matched pair one by one with ***more*** involved legs of children with CP

*** Mann-Whitney U test was used to compare all variables between children with CP and TD

† Number of legs with different strike patterns. R is Rearfoot, M is Midfoot and F is Forefoot

r effect size (r = $z/\sqrt{N}$) and ## The probability that a randomly selected subjects from the CP group would be higher than a randomly selected TD subjects.

2.5 Discussion

The hypothesis that children with diplegic CP would have higher impact peak and load rates during early phase of running, compared to children with TD was supported by the results. Higher impact peak and load rates during early stance phase of running in children with CP, compared to children with TD, which was consistent with Orlin and Laibsirinon’s running study in children with CP (Orlin & Laibsirinon, 2010). High impact forces and load rates in adult runners without neuromuscular conditions have been widely reported to correlate with injuries of bones and muscles around knee and ankle joints, especially stress fracture, patellofemoral pain and other overuse running injuries (Zadpoor & Nikooyan, 2011; Zifchock, Davis, Higginson, McCaw, & Royer, 2008). Abnormally high impact peak and high load rates in children with CP suggested that children with CP may be unable to attenuate excessive load, and therefore may be at higher risk for lower extremity injuries. Existing evidence suggests that the inability to attenuate impact force and load rates may be attributed to a stiff knee joint (Milner, Hamill, & Davis, 2007; Wang et al., 2015). However, increased knee joint stiffness was not observed in children with CP, when compared with children TD. Another contributing factor to excessive high impact force and load rates may be poor adaptation of knee flexion angle caused by excessive knee muscle cocontraction. The authors of this study hypothesized that excessive muscle cocontraction may be present in children with CP during running. Unfortunately, the role of knee muscle cocontraction remains unclear and was not resolved in this study because of unavailable EMG data from the “Run for Life” database. Knee flexion angle during early stance phase of running in children with CP still needs further investigation since no significant differences in the knee contact angle were found between the two groups.

Over similar running distance, higher impact peak and load rates found in children with CP suggested that children with CP could experience higher doses of loading than those with TD. The children with CP (running speed 1.8 m/s) in this study ran much slower than adult runners without neuromuscular conditions (approximately 3.5-3.75 m/s) but average load rates of children with CP were twice as high as those adult subjects (Zadpoor & Nikooyan, 2011). Higher load rates and impact peaks with slower running velocity suggested that legs of children with CP collided with the ground harder and more rapidly than children with TD and those adult runners. More rapid collision between legs and the ground can result in increased stress and micro-trauma to bones, cartilages and other soft tissues (Nigg et al., 1995). If children with CP are able to run faster, higher running speed can contribute to even greater impact force and higher load rates.

None of the other variables of interest were significantly different between children with CP and children with TD. This finding may be attributed to several factors. The Gait Deviation Index (GDI) (Schwartz & Rozumalski, 2008) calculated from walking trials may not be appropriate to classify leg involvement of children with CP during running. The GDI was used for this study because to date, there have been no other methods to classify leg involvement based on running. Leg involvement as determined by preferred foot for hopping and kicking a ball (Maltais et al., 2004), in which leg experiences similar level of load with running, may be considered for future studies. High variability may also have been related to small sample size, heterogeneity of children with diplegic CP and a mixture of strike pattern. High variabilities (see Table 2.2) of all variables in children with TD and CP were also noted. This high variability informs all professionals working with children with CP that careful examination to identify risk for lower extremities injuries due to running or any possible interventions should be individualized, rather than generic intervention programs. Neither clinicians nor parents wants to prevent children from running if they desire and are able to run. Running may be therapeutic for children with CP. Instead, close and follow-up monitoring system and facilitation of self-awareness to detect any muscular discomfort or pain during and after running in children with CP is more critical.

Two main implications for rehabilitation from the key findings of this study are as follows. First, the atypically large impact peak and load rates for children with CP during early stance phase of running may be used to identify those children with CP who may be at high risk for lower extremities injuries. However, risk and causes of running related injuries are multifactorial in nature (Hreljac, 2005). Based on existing evidence, risk factors may be placed in three categories which include personal characteristics (e.g. gender, age and body mass index), training errors, anatomic and biomechanical factors (Hreljac, 2005; Magness, Ambegaonkar, Jones, & Caswell, 2011; Wen, 2007). Impact peak and load rates are parts of biomechanical factors that may be used to identify injuries. Therefore, other factors, such as atypical alignment of lower extremity, have to be considered before identifying children with CP who may be at high risk for injuries. Distance, duration, and cadence for a desired distance of running should be considered with high impact peak and load rates when determining risk for injuries in this population.

Second, the atypical high impact peak and load rates may have implications for the planning of intervention to help children with CP, who desire to run, to run safely. Children with CP recruited in this study were ambulatory routinely without using any assistive devices and did not wear shoes during data collection. However, custom foot and ankle-foot orthotics, and footwear that control proper foot alignment, provide stability, and attenuate increased load, may be beneficial for children with diplegic CP. Positive results using these interventions have been reported in adolescent and adult runners without neuro-musculoskelatal problems (MacLean, Davis, & Hamill, 2008; McMillan & Payne, 2008; O'Leary, Vorpahl, & Heiderscheit, 2008) but not yet investigated in children with CP which could be another interesting area of investigation. Running retraining using visual and auditory feedback has been reported to be effective in decreasing impact forces and load rates in injured runners without neuromuscular conditions (Agresta & Brown, 2015). Running retraining may be a possible intervention for children with CP who have intact or mild impaired cognitive level. A use of visual and auditory feedback has been reported to improve speed and increase stride length during walking in children with CP (Baram, 2012), indicating possibility of running retraining using those feedbacks in order to minimize impact peak and load rates in children with CP, but needs to be investigated.

2.6 Limitations of the Study and Recommended Future Studies

A few limitations of this study should be considered before applying the findings in practice. The limited sample size due to the heterogeneity of the population of children with CP was an obvious limitation of this study. The small sample size limited the generalizability of the findings. However, impact peak and load rates were found significantly different between children with TD and CP, despite the small group and high variability. Non-different results of other research variables may be attributed to heterogeneous group of CP and a mixture of strike patterns. Based on the high inter-subject variability, as evidenced by wide spread of range of all variables of interest between two groups, larger sample sizes are recommended. This would provide a more representative sample with a more stable average. Differences in foot strike pattern may also confound results. Children with TD were included from the unpublished database, to compare with children with CP collected for this study. To resolve this, a stratified sampling based on strike pattern could be a better appropriate design with larger sample size for future studies. However, recruiting children with CP who are able to run may be challenging because they do not often receive therapy services at hospitals or rehabilitation centers. This is because they often have fewer musculoskelatal issues than those who cannot run. Thus, larger sample sizes may be more difficult to achieve. Another limitation is that in the data of children with TD from the unpublished database, EMG data, which were used for CCI, were not available. This missing CCI in children with TD made the role of CCI in children with CP unclear. Other than the biomechanical factors investigated in this study, anatomical variations of lower extremities such as large knee valgus and subtalar overpronation, have been reported to play a role in a development of running related injuries in runners without neuromuscular condition (Saragiotto et al., 2014). Some abnormal alignments of lower extremity, which are commonly seen in children with CP such as excessive hip anteversion and tibial torsion, excessive hip internal rotation, large knee valgus and foot deformity (Morrell et al., 2002), should also be investigated for any potential role in running related injuries when coupled with the high impact variables.

2.7 Conclusion

The important finding of this study is that children with ambulatory diplegic CP had atypically high impact peaks and load rates during early stance phase of running despite slow running speed. These atypically high impact peaks and load rates can be excessive and potential risk factors for the development of musculoskeletal injuries of lower extremities. Although no difference of knee stiffness and knee flexion angle were found between the two groups, their roles on force attenuation mechanism in children with CP remains unclear and needs further investigation, considering the very small sample size in this study. The role of knee muscle cocontraction cannot yet be concluded from this study due to unavailable EMG data and also needs further investigation. Health professionals and parents should keep high impact forces and load rates in mind when encouraging children with CP to run for participation in recreation activities and sport. High impact force and load rates may be used to identify children with CP who are at risk for lower extremity injuries due to running, and may be used to guide potential therapeutic options that minimize excessive impact peak and load rates such as orthoses, footwear, and running retraining.

2.8 References

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CHAPTER 3

Summary and Conclusion

3.1 Modifications to the Research Proposal

This clinical dissertation originally was a preliminary project to fulfil Philosophy of Doctor (PhD) degree in Rehabilitation Sciences program and later became a full clinical dissertation for Doctor of Health Science (DHSc) degree. Thus, a few changes and modifications were made from original PhD proposal to this clinical dissertation for DHSc degree.

First, ten children with diplegic cerebral palsy (CP) and ten children with typical development (TD) were proposed but only eight children with TD and seven children with CP were recruited because difficulty in recruiting subjects and limited number of children with CP who were able to independently walk and run without any assistive devices in Thailand. Health care and para-professionals in Thailand are very limited and less advanced than in the United States. Most children with physical disability including CP do not receive routine physical therapy or any type of exercises. Participation in routine exercise or physical activity for children with physical disabilities has not been widely promoted in Thailand. Children with CP in Thailand are more likely to have sedentary lifestyle that may partly contribute to the inability to walk or loss ability to walk.

Second, foot strike pattern was not a part of initial sampling because study of running in children with diplegic is almost novel. Evidence regarding how children with CP strike the ground does not exist and because of limited number of children with CP who can run. However, different strike patterns have major impacts and can alter all main variables of interest of this study. Seven children with CP contacted the ground with three different strike pattern (rearfoot, midfoot and forefoot); whereas, all eight children with TD from this study struck the ground with forefoot. In order to make comparison, children with

CP from this study who contacted the ground with midfoot and rearfoot were matched paired one by one with the children with TD included from the unpublished database that I was granted by the primary investigator. So, data from only two children with TD who struck with forefoot from this study were used to compare with children with CP. Because of this reason, in total, only six pairs between less involved legs of children with CP and legs of children with TD, and five pairs of more involved legs of children with CP and children with TD were statistically analyzed because no more legs of children with TD striking the ground with midfoot and rearfoot were available from the “Run for Life” unpublished database.

Third, knee muscle cocontraction of children with CP and children with TD were not reported in the results and were not discussed. This is because EMG data were not available from the unpublished database. Cocontraction index of only two LICP and two MICP, compared with those of children with TD were available. Therefore, cocontraction index were not computed and statistically analyzed due to significantly low sample size.

Last, this study aimed to study running in children with CP but running speed of both group were quite slow - more like jogging speed. This is because of the instruction they were consistently provided. The instructions were “run at your comfortable speed” and/or “do not speed up”. In the future study, “run as fast as you can” or faster running speed, if this is possible in children with CP, may be provided more observable results of kinetics and knee contact angles between children with CP and children with TD.

3.2 Implications for Rehabilitation and Recommended Future Studies

The atypically large impact peak and load rates for children with CP during early running may have 2 implications for rehabilitation.

First, excessive impact peak and load rates may be used to identify those children with CP who may be at high risk for lower extremities injuries. However, risk factors and causes of running related injuries are multifactorial in nature (Hreljac, 2005). Based on existing evidence, risk factors may be placed in three categories which include personal characteristics (e.g. gender, age and body mass index), training, anatomic and biomechanical factors (Dennis, 2007; Hreljac, 2005; Magness, Ambegaonkar, Jones, & Caswell, 2011). Impact peak and load rates are some biomechanical factors that may be used to identify injuries. Therefore, other factors have to be considered before identifying children with CP who are at high risk for running related injuries. With given a similar distance of running, higher dosage for running in children with CP when compared with children with TD because of higher cadence of running in children with CP. Higher cadence during running have been reported in children with spastic CP (J. Davids, Bagley, & Bryan, 1998). A combination of high dose of running with high impact peak and load rate should be taken into consideration when determining risk for injuries in this group of population. The findings of increased impact peak and load rates during running still needs further investigation with larger sample sizes and stratified sample based on strike pattern. Additionally, the decision of whether or how much children with CP should be engaged in running should not be solely made based on results from this study. Thorough physical examination and careful monitoring system for more severe pain and discomfort of lower extremities before, during and after running should be well planned. Additionally, larger variability in biomechanical data sets of children with CP, compared with children with TD, suggest that this population is very heterogeneous. Any investigation and intervention should be individualized and thorough in practice.

Second, this result may have implication for the planning of intervention to help children with CP, who desire to run, to run safely. Children with CP recruited in this study were ambulatory routinely without using any assistive devices and did not wear shoe during data collection. However, custom foot and ankle-foot orthotics, cushioned insoles, and footwear that control proper foot alignment, provide stability and likely attenuate the increased load observed in this study. They may, therefore, be beneficial for children with diplegic CP when running. Positive results using these interventions have been reported in adolescent and adult runners (MacLean, Davis, & Hamill, 2008; McMillan & Payne, 2008; O'Leary, Vorpahl, & Heiderscheit, 2008). Significant reductions in pain, impact peak, load rates and knee external moment were reported both short and long term. General indications for orthoses to improve walking gait have been published. Some information from the indications may be applied for children with CP to use during running (J. R. Davids, Rowan, & Davis, 2007), however, it needs to be investigated for actual outcomes in children with CP. Recommendation for types of shoe/cushioned insole to minimize impact force and avoid injuries are not conclusive in runners without neurological problems (Fong Yan, Sinclair, Hiller, Wegener, & Smith 2013). Moreover, specific shoes with and without orthoses during running in children CP have not been explored, but could be another interesting area of investigation. Despite a lack of evidence, orthoses inserted in shoes have been often prescribed for children with CP classified at GMFCS level I-II with the goal of maintaining and improving walking. However, conclusive outcomes are still lacking at both impairment and functional levels due to low quality studies (Figueiredo, 2008; Morrisa, Bowersc, Rossc, Stevensd, & Phillips, 2011; Ridgewell, Dobson, Bach, & Baker, 2010). Therefore, to what extent orthoses, cushioned insole and shoes may have positive impact on children with CP for running is still unanswered and should be a research avenue.

Running retraining may be a possible intervention to reduce impact peak and load rate in children with CP who have intact or mild impaired cognitive level. Running retraining using visual and auditory feedback has been reported to be effective in decreasing impact forces and load rates in injured runners without neuromuscular conditions (Agresta & Brown, 2015). Visual and auditory feedback using specifically designed devices to improve walking speed and stride length in children with CP have been investigated (Baram, 2012), indicating possibility to modify running style in children with CP in order to reduce impact peak and load rates. Again, running retraining by using visual and auditory feedback in children CP needs to be investigated.

3.3 Conclusion

Children with ambulatory diplegic CP demonstrated higher impact peak and load rates during early stance phase of running than children with TD although they ran at similar speed. High impact peak and load rates are strongly associated with running related injuries in adult runners without neuromuscular problems. Thus, children with CP may be at a high risk for running related injuries. The finding of non-significant results for knee stiffness, knee flexion angles and knee excursion at early stance running between children with CP and TD were likely attributed to very small sample sizes and the mix of strike pattern. Knee muscle cocontraction during running in children with CP and children with TD were not statistically analyzed in this study because of very small sample size. The role of knee muscle cocontraction on force attenuation mechanism needs further investigation. The findings from this study have important implications. Impact peak and load rates, coupled with thorough physical examination for malalignment of lower extremities, may be used to identify children with CP who are at risk for injuries due to running. These assessments should be combined with the experience of musculoskelatal pain and discomfort during and after running. Therapeutic options such as footwear, orthoses, cushions, and running retraining that targets decreasing impact peak and load rates may aid children with CP in running safely.

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VITA

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EDUCATION

INSTITUTION AND LOCATION	DEGREE	YEARS	FIELD OF STUDY
Mahidol University, Thailand	B.S.	1994-1998	Physical Therapy
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A. Current Positions and Honors

Positions and Employment

2014 – present	Acute care and rehabilitation therapist, Nemours/Alfred I. DuPont Hospital, Wilmington, DE
2013- present	Early intervention physical therapist, Delaware
2013 – 2014	School based physical therapist, Pennsylvania
2003 – 2013	Faculty member, Department of Physical Therapy, Faculty of Allied Health Science, Thammasat University (Thailand)

Other Experience and Professional Memberships

2008 – 2010	**Member,** American Physical Therapy Association (APTA), Pediatric Section
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B. Peer-Reviewed Publications

1. Laibsirinon S, Jarususin N, Manakiettichai T, KoiKoy C. (2012). Pulmonary function and chest expansion in Thai boys with Down syndrome. Thammasat Medical Journal , 12(2), 269-275.
2. Laibsirinon S, Prapetch J, Tonpoo P, MoolMok P. (2012). Comparison between Gross Motor Function Measure-88 and -66 in evaluating gross motor function in Thai infants with typical development. Thammasat Medical Journal, 12(2), 344-352.
3. Laibsirinon S. (2011). Hippotherapy in children with Cerebral Palsy. Thammasat Medical Journal, 11(4), 634-641.
4. Laibsirinon S, Mahasup N, Tantileepikorn P. (2008). Inter-rater reliability of Thai version of Gross Motor Function Classification System (GMFCS) in Thai children with cerebral palsy. Journal of Thai Physical Therapy, 30 (1), 26-35.

D. Research Support

Thammasat University *2011-2012*
Comparison of impact forces, knee position and knee muscle cocontraction during running in children with cerebral palsy and children with typical development
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Thammasat University *2004-2005*
Reliability of Gross Motion Function Classification System in Thai version for children with Cerebral palsy”
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